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Case Report

Pulmonary adiaspiromycosis: The first reported case in Turkey

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ABSTRACT

Adiaspiromycosis is a world wide airborne infection and is a pulmonary disease in humans caused by the fungus *Chrysosporium parvum var crescens (Emmonsia parva)*. It is caused by inhalation of spores of the saprophytic soil fungus. The spherules induce a granulomatous reaction, in which a single spherule is surrounded by a fibronecrotic and suppurative or fibrous granuloma. The disease can be localized and asymptomatic, or disseminated, occasionally severe, or even fatal. Reticulonodular infiltrates are the most common radiographic manifestations, with localized infiltrates occurring occasionally. We present a case with bilateral diffuse interstitial micronodular infiltration and granulomatous inflammation in the lung. The larvae of adiaspiromycosis visualized in the center of granulomatous inflammation by histopathologic examination of the lung biospy specimens. We aimed to discuss the very rare cause of diffuse interstitial and granulomatous inflammation of lung.

1. Introduction

Adiaspiromycosis is a rare fungal infection in the lung and is caused by inhalation of spores of the saprophytic soil fungus Chrysosporium parvum var crescens (previously known as Emmonsia crescens). The organisms are large spherules (mean diameter, >200 μm; range, 50–500 μm) that have trilaminar walls surrounding empty centers.[1,2] As adiaconidia do not duplicate, the lesions are usually limited to the lungs and the symptomatology and radiological findings are directly related to the number of inhaled conidia. Reticulonodular infiltrates are the most common radiographic manifestations, with localized infiltrates occurring occasionally. The disease is usually self-limited, although fatal cases have been reported. A small inoculum produces no or mild clinical symptoms and scattered radiological pathology. But those individuals submitted to an heavy or repeated inoculum may present an acute, sometimes severe, pulmonary disease, with granulomatous lesions distributed randomly throughout both lungs.[3] We aimed to present and discuss of the clinic, radiologic and pathologic features of pulmonary adiaspiromycosis in a patient which the first reported a case in Turkey.

2. Case report

A 62 year-old farmer male was admitted to the hospital with the complaints of weakness and dyspnea for one month. He is ex-smoker during 5 years with the 20 pack/year smoking history. On admission, temperature, cardiac pulse, respiratory rate and blood pressure were 37 °C, 84 beats per minute, 16 per minute and 110/70 mmHg, respectively. Bilateral inspiratory crackles were heard on the lung oscultation. There was no any abnormality on other physical examination. The hemoglobin value was 11.1, peripheral blood leukocyte count was 10900 cell/cu mm and erythrocytes sedimantation rate was 97 mm/h. Arterial blood gas values during room-air breathing revealed that the pH: 7.35, pO2: 80 mmHg, pCO2: 35 mmHg and SaO2:94.4%. Other laboratuary values were normal. Pulmonary function test values showed that FVC was 2440 ml(77% of predicted), FEV1 was 2130 ml(84% of predicted) and FEV1/FVC was 87%. A chest roentgenogram showed the bilateral diffuse interstitial micronodular infiltrates(Fig. 1). Thorax CT revealed the bilateral diffuse peribronchial and interstitial reticulonodular infiltrates(Fig. 2). Bronchoscopy was performed and it was normal. The cytologic examination of bronchial and bronchoalveolar lavage(BAL) were benign. BAL lavage showed 77% alveolar macrophage, 16% lymphocytes and 7% neutrophils. The microscopic pathologic examination of transbronchial lung biopsy was non-diagnostic. The lung biopsy with video-assisted thoracoscopc surgery(VATS) was performed. In the histopathologic microscopic examination, the granulomas with parasite-like

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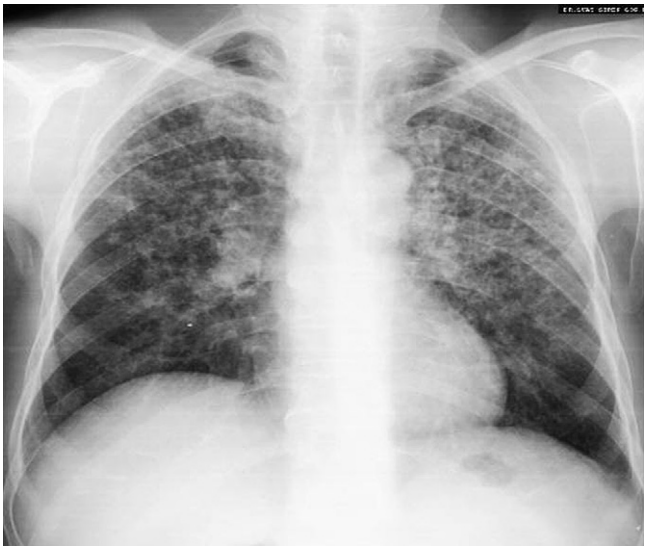

Fig. 1. Chest roentgenogram is showing bilateral reticular and onodular infiltrations.

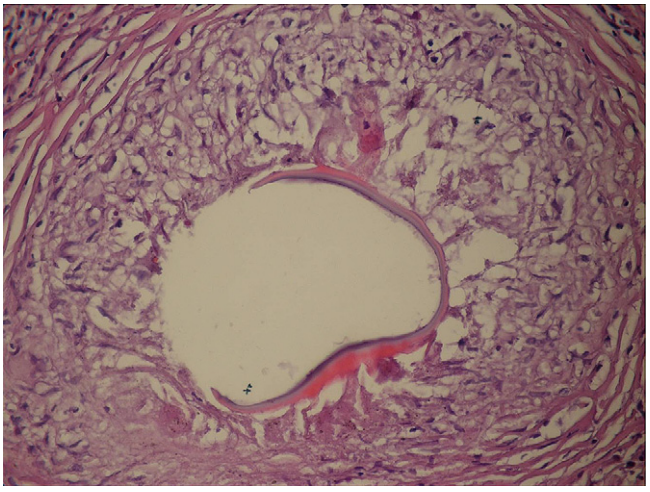

Fig. 3. Histopathologic image(H&E × 400) showing a well shaped Aconodium larva in the center of granulomatous inflammation.

structures were seen in the lung biopsies. The patient was treated for three weeks with oral Albendazole 800 mg/day. After the treatment, a slight decrease in the pulmonary infiltration was seen on chest roentgenogram, but there was no fully recorvered. The pathologic specimens were consulted to a microbiologist; special stains for fungi were done which revealed the characteristic adiaconidia. The larvae of adiaspiromycosis visualized in the center of granulomatous inflammation with H&E and Grocott-methenamine silver stains(Figs. 3,4 and 5). The patient had followed without any treatment and the symptoms and radiologic abnormalities almost fully improved after seven months(Fig. 6). The approval of patient and institution were taken to use their records for our study.

3. Discussion

Adiaspiromycosis is a world wide airborne infection and is a pulmonary disease in humans caused by the fungus *Chrysosporium parvum var crescens (Emmonsia parva)*. It is essentially restricted to the lungs. The spherules induce a granulomatous reaction, in which a single spherule is surrounded by a fibronecrotic and suppurative or fibrous granuloma. The disease can be localized and asymptomatic, or disseminated, occasionally severe, or even fatal. The severity and extent of the disease in the lungs depends on the amount of the dustborne conidia inhaled.[1,3,4]

The disease was firstly described by Emmons and Ashburn in Arizona rodents in 1942, and the first case with adiaspiromycosis was reported in France in 1964. Several asymptomatic cases were reported like in the first case.[5,6] In 1971,Kodousek et al. reported a symptomatic adiaspiromycosis case with respiratory infection

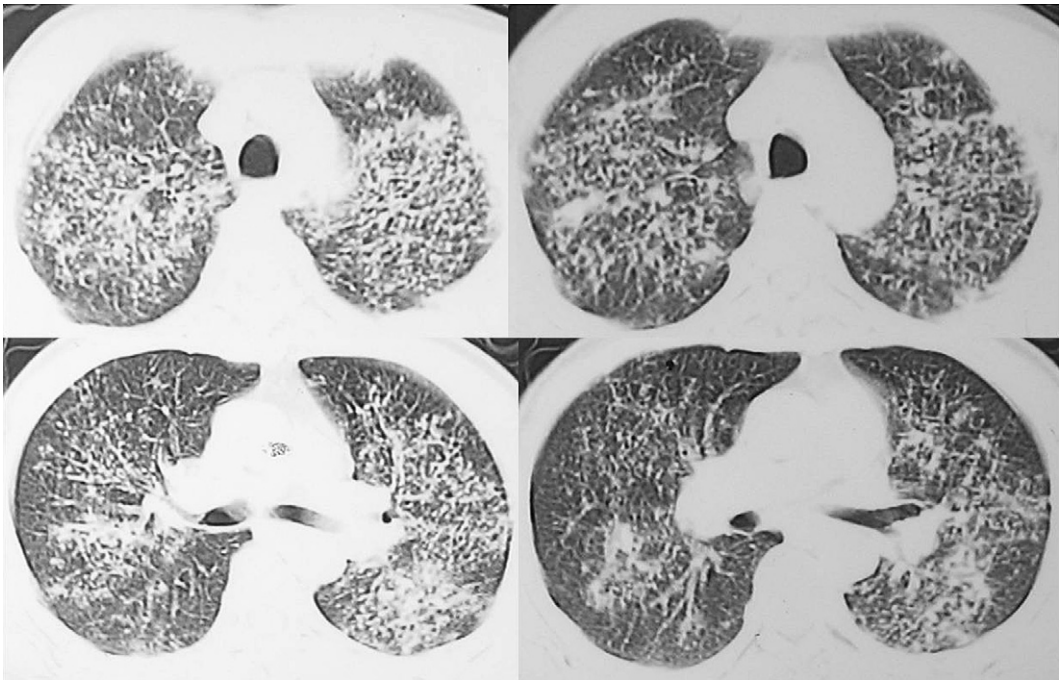

Fig. 2. Thorax CT scans are showing the bilateral interstitial and peribronchial reticulonodular infiltrations.

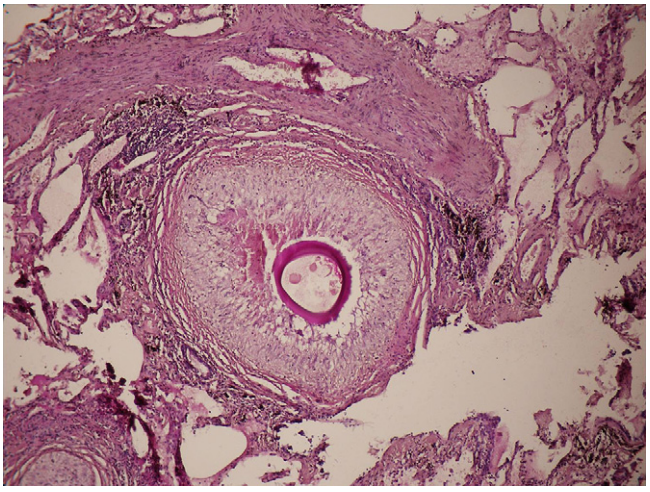

Fig. 4. Histopathologic image(PAS × 100) showing the periodic acid Schiff(PAS) positive Aconidium larva in the granulomatous inflamation.

findings and diffuse bilateral pulmonary lesions, as seen in our patient.[7]

To our knowledge, approximately thirty patients published in literature. The range of age was 2–74 years old. Only two of them were female. Interestingly, the youngest and oldest patients were female. Adiaspiromycosis is usually reported in males, as seen in our patient.

Diffuse interstitial reticulonodular infiltrates are the most common radiographic findings in reported symptomatic cases with aidaspiromycosis. However, asymptomatic cases usually tend to be localized and diagnosed by incidentally. The symptoms and radiologic involvement of the disease in the lungs depends on the amount of the dustborne conidia inhaled. There are three types of adiaspiromycosis; *1.Simple pulmonary adiaspiromycosis*; is an incidental findings in lung tissue removed for another reason and the patients with this form of the disease have no symptoms or radiographic abnormalities attributable to the fungal infection. *2.Disseminated pulmonary adiaspiromycosis;* localized or diffuse bilateral pulmonary disease that as a finely nodular reticulonodular radiographic pattern; only those patients with the densest, most severe bilateral involvement have had symptom, which include fever, cough and progressive dyspnea, as seen in our patient. The spontaneous remission was reported in these form.[8] *3.Fulminan pulmonary adiaspiromycosis;* disseminated pulmonary adiaspiromycosis can progress to the respiratory failure and death. We called as "fulminan pulmonary adiaspiromycosis" according to reported articles.[2,9]

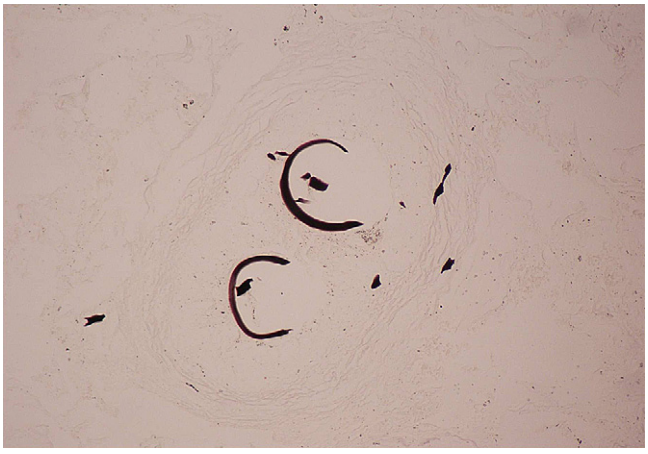

Fig. 5. The larvae of adiaspiromycosis visualized with Grocott-methenamine silver stain(Grocott × 400).

Fig. 6. The almost fully recorvery is showing on chest roentgenogram at 7th months.

The reported symptoms were nonspesific and acute or subacute onset. The clinic, radiologic and histopathologic findings are very similar to pulmonary tuberculosis, sarcoidosis, histoplasmosis. We suggested that, the clinic, radiologic and histopathologic findings are not only caused from infectious ability of aidiasporoconidium. Also, Type IV hypersensitivity reaction againts to aidiasporoconidium may contribute to the clinic, radiologic and histopathologic findings.

The diagnosis of pulmonary adiaspiromycosis is difficult. It may also be kept in mind especially in patients who have diffuse interstitial reticulonodular infiltrates and granulomatous inflammation in histopathologic examination. Adiaconidia are not seen or isolated in culture from sputum or bronchoalveolar fluid, because they do not multiply and remain trapped within lung granulomas, as in our patient. The diagnosis is usually achieved by chance, in specimens of biopsied lung tissue stained routinely by H&E. In literature, the reported cases were diagnosed with histopathologically which achived by open lung biopsy, transbronchial biopsy or autopsy.[3] Histological examination of the lungs demonstrated a large number of, 100–200 mm large, thick-walled spores surrounded by a chronic granulomatous inflammatory reaction with presence of lymphocytes, macrophages, giant cells and connective tissue (Fig. 1). The spores were positively stained with Grocott stain and were identified as the fungi *Chrysosporium parvum* var. *crescens* (*Haplosporangium parvum* and *Emmonsia parva*).[11,12]

The drug treatment of adiaspiromycosis is still controversial. There have been reports of the use of monotherapy or combinations of amphotericin B, 5-fluorocytosine, ketoconazole, itraconazole, and systemic corticosteroids in pulmonary adiaspiromycosis. The spontaneous remissions without any treatment were reported in the patients with disseminated pulmonary adiaspiromycosis.[10] In our case, the treatment started with the Albendazole and three weeks later, minimal regression in reticulonodular infiltrations was observed on chest roentgenogram. The histopathologic specimens were consulted with microbiologist and pathologist due to slight regression of disease, the adiaspiroconidium was confirmed. After that, we did not give any drugsand radiologic abnormalities with sypmtoms were almost fully improved after seven months.

In conclussion; It is the first case report in Turkey with disseminated pulmonary aidaspiromycosis. It should be kept in mind for diagnosis of adiaspiromycosis in patients who have

pulmonary reticulonodular infiltrations and granulomatous inflammation in the lungs.

Conflict of interests

None declared.

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